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Successful treatment for intra-abdominal bleeding due to spontaneous rupture of huge liver cyst using transcatheter arterial embolization: a case report

Koichiro Mitsuoka, Yosuke Miyachi, Shuntaro Hirose, Takashi Taketa, Akihiro Suzuki, Taketo Matsubara, Tadao Yokoi, Gen Shimada, Akihiro Kishida, Toshimi Kaido

Department of Gastroenterological and General Surgery, St Luke's International Hospital, 9-1 Akashi-cho, Chuo-ku, Tokyo, Japan

ABSTRACT

Background: Non-parasitic simple liver cysts are one of the most common benign hepatic lesions. Although most liver cysts are asymptomatic and remain silent throughout the patient's life, extremely large cysts can become symptomatic by direct compression to adjacent organs. Herein, we report a case of a spontaneously ruptured simple liver cyst, which is a rare presentation of a benign liver cyst. The patient's liver cyst re-ruptured and was treated with transcatheter arterial embolization (TAE). **Case report:** A 62-year-old man presented to our hospital complaining of acute-onset lower abdominal pain. He had undergone laparoscopic fenestration of a huge liver cyst in another hospital 2 years prior. Computed tomography (CT) scan showed spontaneous rupture of a large liver cyst. Laparoscopic exploratory laparotomy showed no signs of ongoing intra-abdominal bleeding from the liver cyst; therefore, the operation was completed with peritoneal lavage. The patient was discharged from our hospital on postoperative day 5. Twelve days after the initial presentation, the patient was re-admitted to our hospital complaining of recurrence of lower abdominal pain. CT scan showed an enlargement of the previously ruptured liver cyst, with intra-abdominal bleeding and massive hematoma in the cyst. Extravasation of the cyst's wall was also detected. Under the diagnosis of intra-abdominal bleeding from the artery in the wall of the huge cyst, emergent TAE was performed. Although the exact spot of extravasation was not detected, the anterior segment branch of the right hepatic artery, which corresponds to extravasation shown on the CT scan, was embolized. The patient was discharged from our hospital after 7 days, and the liver cyst remained stable without abdominal pain for more than 2 months. **Conclusions:** This case highlights a rare presentation of spontaneous rupture of a liver cyst with massive bleeding and the efficacy of TAE for the conservative treatment of ruptured liver cysts.

Keywords: liver cyst, massive bleeding, rupture, lower abdominal pain, transcatheter arterial embolization, right hepatic artery, extravasation

*Correspondence to Author:

Toshimi Kaido

Department of Gastroenterological and General Surgery, St Luke's International Hospital, 9-1 Akashi-cho, Chuo-ku, Tokyo, Japan

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Background

Non-parasitic simple liver cysts are one of the most common benign hepatic lesions. The prevalence of liver cysts has been increasing due to the development of diagnostic imaging techniques such as computed tomography (CT). Most liver cysts are asymptomatic and remain silent throughout the patient's life. Symptoms related to liver cysts, although very rare, are usually elicited by extremely large cysts directly compressing their adjacent structures, such as the stomach, duodenum, or, in some case reports, inferior vena cava, which leads to heart failure [1-2]. On the other hand, spontaneous rupture, sometimes seen in hepatocellular carcinoma or other benign hepatic lesions such as hydatid cysts or hepatocellular adenomas, is extremely rare in liver cysts.

Here, we present the case of a 62-year-old man who was admitted to our hospital with spontaneous rupture and re-rupture of a liver cyst. The patient was successfully treated with transcatheter arterial embolization (TAE) after a second spontaneous rupture.

Case report

A 62-year-old man presented to our hospital complaining acute-onset lower abdominal pain. His past medical history was remarkable for the fenestration and omentum patch for multiple liver cysts (S2, S5 and S7/8) in another hospital 2 years prior. Physical examination revealed rebound tenderness in the lower abdomen. The result of CT scan showed multiple large irregular liver cysts in the right lobe. Intraperitoneal fluid adjacent to the cysts was also noted; therefore,

spontaneous rupture of the liver cyst was suspected (Fig. 1a). Deep venous thrombosis and pulmonary artery embolism (DVT/PE) were incidentally noted on the same CT scan. Thus, laparoscopic exploratory laparotomy was urgently performed. The surface of ruptured liver cyst was covered with greater omentum; however, it was unclear whether the cysts were same as the previously fenestrated ones. Although a small amount of bloody ascites was noted, there were no signs of ongoing intra-abdominal bleeding from the liver cyst; therefore, the operation was completed with peritoneal lavage (Fig. 2). After the operation, oral rivaroxaban was started for DVT/PE, and the patient was discharged from our hospital on postoperative day 5.

Twelve days after the initial presentation, the patient was re-admitted to our hospital complaining of recurrence of lower abdominal pain. Other than mild low blood pressure (94/66 mmHg), his vital signs were within the normal range at presentation. Physical examination revealed a flat soft abdomen without tenderness. Blood tests also showed no signs of anemia (Hemoglobin 12.3 g/dL). Furthermore, CT scan showed an enlargement of the previously ruptured liver cyst (Fig. 1b). A large hematoma (indicated by an arrow) and extravasation at the wall of the cyst (indicated by an arrowhead) are also noted. Under the diagnosis of re-ruptured liver cyst, emergent TAE was performed. Although the exact spot of extravasation was not detected at angiography, the anterior segment branches of the right hepatic artery, which

corresponds to extravasation shown in the CT scan, were embolized (Fig. 3a-d). The right gastroepiploic artery ran close to the extravasation; therefore, the artery was also examined during TAE. The result showed no extravasation from the artery, and it was left untreated. The patient was discharged from our hospital after 7 days. Oral rivaroxaban, which had been discontinued during the second

admission, was restarted after discharge. On outpatient department, the liver cyst was evaluated with magnetic resonance imaging (MRI) and there were no abnormal findings such as nodule formation or thickening of the wall, as well as further enlargement or new hemorrhage (Fig. 4a&b). The liver cyst remained stable for more than 2 months without recurrence of abdominal pain (Fig. 1c).



Fig. 1 (a) The ruptured liver cyst at first presentation. The large liver cyst was noted on the right lobe of the patient's liver. (b) The result of CT scan at second presentation. Note the enlargement of the previously ruptured liver cyst. The arrow indicates the hematoma inside the liver cyst. The extravasation from the liver cyst wall is indicated by the arrowhead. (c) The result of follow up CT scan obtained 2 months after final admission. The size of the hematoma inside the liver cyst did not increase.



Fig. 2 The operational finding of the ruptured liver cyst at first presentation. Although a small amount of bloody ascites was noted, there were no signs of ongoing intra-abdominal bleeding from the liver cyst.

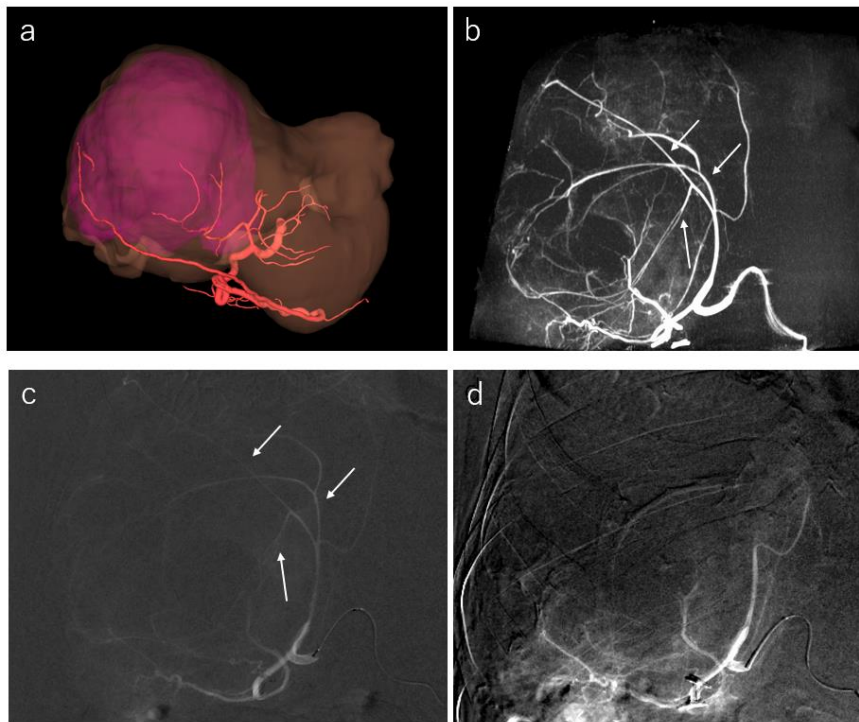


Fig. 3 (a) The result of vascular image reconstruction using SYNAPSE VINCENT V5.5.0007 at second presentation. (b&c&d) The result of transcatheter arterial embolization. Note that there are no obvious extravasation nor pseudo-aneurysm. The anterior segment branches of the right hepatic artery, which corresponds to the extravasation shown in the CT scan (indicated by arrows in Fig. 3b&c), were embolized.

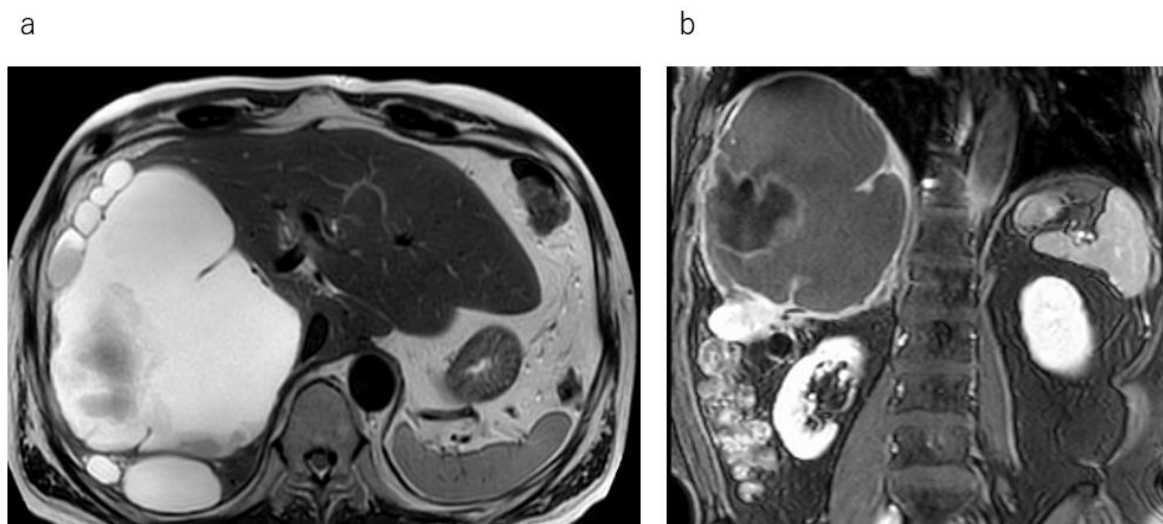


Fig. 4 (a, b) MRI results. There were no signs of possible cystadenoma or cystadenocarcinoma.

Discussion

Spontaneous rupture of a liver cyst is extremely rare; therefore, it currently has no specific

treatment strategy to date. And there are no reports on what imaging features predict spontaneous rupture or bleeding. Although

several authors have reported their experiences with surgical treatments, including cystectomy or hepatectomy for liver cyst rupture [3-6], these may be too invasive to be performed in an emergency setting. Compared with highly invasive treatment options, TAE is minimally invasive and has a high possibility of preventing re-bleeding. As shown in this case, re-rupture of the hepatic cyst can sometimes occur, especially when the patient is on anticoagulation therapy. Although anticoagulation therapy might induce rupture and re-rupture from the huge liver cyst, simple observation is insufficient for the treatment of liver cyst rupture. Therefore, it is necessary to take measures to prevent re-rupture and intra-abdominal bleeding from the cystic wall, and TAE may be an effective treatment method.

In this case, the exact location of extravasation was confirmed by CT scan; therefore, although the bleeding site could not be identified during TAE, embolization was successfully completed. It seems that the elevation of intra-cystic pressure caused spontaneous hemostasis while waiting for TAE; however, as previously shown, this does not mean that re-bleeding could be prevented fully. In such cases, prophylactic embolization based on the results of CT scans may be effective in preventing re-bleeding. Although Ishikawa et al. [7] demonstrated a case of an enlarged liver cyst that ruptured again after TAE, we should avoid concluding that TAE is not suitable for the prevention of re-rupture of liver cysts. Because they did not mention the details of TAE, it is unclear whether they could identify the exact bleeding site during TAE. In addition,

they reported that percutaneous puncture and aspiration of the ruptured cyst were performed after TAE, and this might have decreased the intra-cystic pressure and caused re-bleeding if the exact source of bleeding was left untreated. In our case, we were able to identify the blood vessel that caused the liver cyst rupture by CT scan and embolized it prophylactically. Although the observational period was still several months, the cyst did not grow afterwards.

Conservative therapy using TAE for the spontaneous rupture of liver cysts inevitably has some drawbacks. There have been some reports of spontaneously ruptured cystadenoma and cystadenocarcinoma [5], and it is very important to differentiate these conditions when treating them conservatively with TAE alone. Although there are no specific imaging features that permit the reliable differentiation of cystadenoma from cystadenocarcinoma, both are usually seen as multilocular cystic lesions with septa or solid nodules that are sometimes accompanied by calcification; therefore, the differentiation between simple liver cyst and cystadenoma or cystadenocarcinoma is not very difficult [2, 8]. If these conditions are suspected on imaging findings, additional procedures such as resection of the cyst may be needed. Because there were no such features that suspected the possibility of cystadenoma or cystadenocarcinoma in this case (Fig. 4a&b), the patient was kept under observation. In addition, the long-term outcomes after TAE remain unclear. The observation period after TAE in this case is still several months; therefore,

the patient's condition should be continuously evaluated.

Conclusions

We report a case of spontaneous rupture of a liver cyst with massive bleeding. TAE is minimally invasive and very effective for hemostasis and prevention of rebleeding.

Declarations

Ethics approval and consent to participate

Not applicable.

Consent for publication

Not applicable.

Availability of data and materials

The datasets used and/or analyzed in the present study are available from the corresponding author upon reasonable request.

Competing interests

All authors declared no conflict of interests.

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Authors' contributions

M.K. contributed to the design and implementation of the research, analysis of the results, and writing of the manuscript.

M.Y. and K.T. made substantial contributions to the conception or design of the work, or the acquisition, analysis, or interpretation of data for the work, revising it critically for important intellectual content, and final approval of the version to be published.

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Author's information

Department of Gastroenterological Surgery, St. Luke hospital, 9-1 Akashicho, Tokyo, Japan

List of abbreviations

CT: computed tomography; DVT : Deep venous thrombosis; MRI : magnetic resonance imaging; PE : pulmonary artery embolism; TAE: transcatheter arterial embolization

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